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Clinically Small Tonsils Are Typically Not Obstructive in Children During Drug-Induced Sleep Endoscopy

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Abstract

Objective—To determine whether the degree of lateral pharyngeal wall (LPW) obstruction on pediatric drug-induced sleep endoscopy (DISE) correlates with preprocedure tonsillar hypertrophy score on physical examination, and to determine if clinically small tonsils are obstructive.

Methods—Retrospective review of 154 patients who underwent DISE at a single pediatric tertiary care center over a 4-year period. Inclusion criteria were documentation of Brodsky tonsil score on preoperative physical examination. Exclusion criteria were previous tonsillectomy, adenoidectomy, or adenotonsillectomy. Lateral pharyngeal wall obstruction was graded for each patient from 0 (no obstruction) to 3 (severe obstruction) using a validated pediatric DISE scoring system known as the Chan-Parikh scoring system (C-P). Data were analyzed using multivariate linear regression controlling for age at time of DISE and presence of comorbid conditions.

Results—One hundred fifteen patients met criteria for analysis. Median age at DISE was 5.1 years. A moderate positive correlation was calculated between Brodsky score and DISE score, Spearman correlation coefficient 0.55, $P = < 0.001$. Linear regression modeling determined that for every 1-point increase in tonsil score, there was a 0.7-point increase in C-P LPW score (95% confidence interval [0.45, 0.92]). Sensitivity analysis did not detect a difference in correlation between children with comorbid conditions and children who were otherwise in good health. Of the 65 children with a pre-DISE Brodsky tonsil score of 1, 39 (60%) had a LPW score of 0 (no obstruction); nine (14%) had a score of 1 (< 50% obstruction); 11 (17%) had a score of 2 (> 50% obstruction); and six (9%) had a score of 3 (100% obstruction).

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Conclusion—There is a positive correlation between Brodsky Score and DISE LPW score. The majority of children with sleep-disordered breathing with a Brodsky score of 1 did not demonstrate LPW obstruction. These children may benefit from DISE for identification of alternative sites of upper airway obstruction.

Keywords

Obstructive sleep apnea; drug-induced sleep endoscopy; polysomnogram; sleep-disordered breathing; pediatrics

INTRODUCTION

Obstructive sleep apnea (OSA) affects 1% to 4% of children, and has been linked to a number of health-related issues, such as behavioral problems, growth failure, cor pulmonale, and other morbidities.[1–3] Children with OSA have been found to have decreased health-related quality of life.[4] Adenotonsillectomy is the recommended surgical treatment for children with OSA,[5] and randomized-controlled trials have found that adenotonsillectomy generally improves symptoms and quality of life of children with OSA.[6]

However, children with small tonsils may not experience the same benefit from adenotonsillectomy. Tonsil size is commonly rated using the Brodsky standardized system for evaluation of tonsil size (Fig. 1).[7] It is possible that children with small tonsils may have other sources of obstruction contributing to their OSA, which could lead to residual disease. Imanguli and Ulualp found that children with grade I tonsils had a median postoperative Apnea-Hyponea (AHI) of 2.2 after adenotonsillectomy, indicating that the majority of patients had residual OSA despite surgery.[8]

Studies have shown up to 30% of pediatric patients will have residual OSA after adenotonsillectomy.[9] This observation has led to increased interest in the role of drug-induced sleep endoscopy (DISE) in the evaluation of airway obstruction in children with OSA. DISE typically involves a flexible fiberoptic examination of the upper airway performed when the child is under sedation while maintaining spontaneous ventilation.[10,11]

We have previously described and validated a systematic approach to scoring severity of airway obstruction at five different levels during DISE, including the region of tonsillar obstruction: the lateral pharyngeal wall (Fig. 1).[12] The objective of this study was to compare preoperative Brodsky tonsil score to lateral pharyngeal wall obstruction diagnosed during DISE. If patients with small tonsils do not display significant obstruction during DISE, evaluation for other sites of obstruction in children with small tonsils may be warranted.

MATERIALS AND METHODS

This study was based at Seattle Children's Hospital, Seattle, Washington, a pediatric tertiary care facility. Institutional review board approval was obtained prior to data collection. At Seattle Children's Hospital, surgeons performing DISE rate the severity of obstruction in a standardized fashion using the Chan-Parikh (C-P) scoring system. This score is based on the assessment of five anatomic locations: adenoid, velum, lateral pharyngeal wall, tongue base, and supraglottis. Each site is graded on a 4-point scale based on severity of obstruction: 0 = no obstruction, 1 = < 50% obstruction, 2 = > 50% obstruction, and 3 = complete obstruction. The anesthetic technique for all DISE utilized sevoflurane and propofol per institutional protocols. Sleep endoscopy scores were noted at the time of surgery by the surgeon responsible for each case. Records are then stored in an institutional database, along with demographic characteristics and polysomnography (PSG) parameters.

Records of all patients in the database were screened between January 1, 2011, and March 30, 2016. All patients who had undergone DISE and had an available preoperative Brodsky tonsil score were included in the study. The decision to perform DISE was based on clinical evaluation by the individual attending surgeon. DISE was more commonly performed on children perceived to be at greater risk for residual OSA, such as those with neurological disorders, syndromes such as trisomy 21, small tonsils, or with clinical suspicion for multilevel airway obstruction. Children who had already undergone adenotonsillectomy, adenoidectomy, or tonsillectomy were excluded from the study. Children were stratified into groups based on the presence or absence of significant comorbid conditions. Significant comorbid conditions were defined as those conditions that could affect muscle tone, adversely affect neurologic status, or produce anatomic abnormalities associated with upper airway obstruction. Common conditions include trisomy 21, cerebral palsy, and Pierre-Robin sequence.

The majority of the subjects had undergone PSG prior to DISE. Subjects underwent PSG at an accredited sleep laboratory as part of clinically indicated care, with results interpreted by board-certified pediatric sleep medicine physicians. Polysomnograms were scored in accordance with American Academy of Sleep Medicine parameters.[13] Apnea-Hypopnea index and lowest recorded oxygen saturation were noted from the preprocedural PSG.

Analysis

Children were grouped into categories based upon Brodsky score and LPW score on endoscopy. Univariate analyses were performed to obtain descriptive statistics for each group. Means were calculated for continuous variables such as AHI and average age at time of DISE. Proportions were calculated for binary variables such as presence of syndrome. Spearman correlation coefficient was calculated for association between Brodsky and LPW score. A multivariate linear regression model was created, controlling for age at time of DISE and presence of comorbid conditions. Sensitivity analysis was performed to determine if there was a significant difference in association between children with comorbid conditions and those without. Additional analysis was performed on each cohort to compare tonsil size and OSA severity.

For all tests, $P < 0.05$ was considered statistically significant. Stata 13.1 (Stata Inc., College Station, TX) statistical software was used for all analyses.

RESULTS

There were 154 children with DISE results; of those, 115 children had both preoperative Brodsky score and LPW score. Table I provides patient characteristics and PSG results for the children based on Brodsky tonsil score and LPW score. Median age at DISE was 5.1 years. More than half of the children had grade I tonsils, and the majority of them had a LPW score of 0. Figure 2 and Figure 3 are scatter plots depicting the association between AHI score and Brodsky tonsil score and LPW score. Spearman correlation analysis identified similar but weak positive correlation coefficients between tonsil size and AHI score, $r = 0.23$, $P = 0.02$, and LPW score and AHI score, $r = 0.23$, $P = 0.02$. There was not a statistically significant correlation between oxygen nadir and either tonsil size or LPW score.

Of the included patients, 51 of the 115 had significant comorbid conditions that potentially could contribute to OSA. When we limited our analysis to children with these conditions, we found no correlation between tonsil size and AHI score ($r = -0.06$, $P = 0.7$), indicating that the source of their upper airway obstruction may be elsewhere. Among children without these conditions, we found a moderate correlation between tonsil size and AHI score ($r = 0.4$, $P = 0.002$) (Fig. 4a and 4b).

We then looked at the association between Brodsky and LPW scores. Figure 5 is a weighted scatter plot depicting the relationship between the two measurements. Of the 65 children with a Brodsky score of 1, 39 (60%) had a LPW score of 0. A moderate positive correlation was calculated between Brodsky score and LPW score, Spearman correlation coefficient 0.55, $P = < 0.001$.

A multivariate linear regression model was then generated controlling for age at time of endoscopy and presence of syndrome. Linear regression modeling determined that for every 1-point increase in tonsil score, there was a 0.7-point increase in LPW score (95% confidence interval [0.45, 0.92]) (Fig. 6).

Sensitivity analysis did not detect a difference in correlation of tonsil size and LPW obstruction between children with comorbid conditions and children who were otherwise in good health. Spearman correlation coefficient for children with comorbid conditions was 0.53, which decreased slightly to 0.44 among children without comorbid factors; however, Fisher r-to-z transformation did not find this difference to be significant, z = 0.76, $P = 0.45$.

Of the patients with 1+ tonsil score, the average adenoid score was 1.22 versus the average in the entire cohort of 1.24. Chan-Parikh score at other sites is included in Table II.

DISCUSSION

Obstructive sleep apnea in children is a recognized contributor to poor quality of life and disturbance in development.[1] The American Academy of Pediatric guidelines currently recommend adenotonsillectomy as first-line therapy for children with OSA in patients with adenotonsillar hypertrophy.[14,15] Yet, controversy exists as to whether tonsillectomy should be routinely performed on children with OSA regardless of tonsil size.

In healthy children with OSA, small tonsils and adenoids may not be completely responsible for their obstructive symptoms. In fact, previous studies have shown that tonsil size does not always correlate well with baseline OSA severity, even when adenoid hypertrophy is incorporated into the analysis.[16] Many of these studies excluded patients with other comorbidities and syndromes. In our analysis, we demonstrated a positive correlation between tonsil size and LPW collapse on DISE. Performing a subgroup analysis, we found that tonsil size did correlate with OSA severity among otherwise healthy children without comorbid condition, but we did not find a similar relationship among children with comorbid conditions contributing to OSA. Causes for these persistent and complex cases of OSA are numerous and include hypotonia, craniofacial disproportion, lingual tonsillar hypertrophy, and laryngomalacia, all of which may contribute to OSA despite adenotonsillectomy.

DISE has recently evolved as a diagnostic tool for identifying sites of obstruction in children with OSA.[12] DISE was initially used to assess children with residual OSA who have already undergone adentonsillectomy.[10] However, because more data support the utilization of DISE-directed surgery in pediatric OSA, surgeons may be more inclined to perform DISE on children with comorbid conditions or small tonsils prior to adenotonsillectomy.

In this study, our first aim was to assess for correlation of preoperative assessment of tonsil physical exam score with DISE LPW score. Our second aim was to look specifically at children with small tonsils (Brodsky 1) to see if these were obstructive. We noted a statistically significant positive correlation between Brodsky and LPW scores, and we found that the majority of small tonsils are not obstructive during DISE. No previous studies to our knowledge have examined correlation between tonsil size and LPW collapse on DISE. The association between tonsil size and OSA have been reported previously with varying results.[17] However, conclusions from this study warn against estimations of OSA severity based solely on tonsil size, particularly among children with comorbid conditions. Our findings, specifically that small tonsils generally are not obstructive on DISE, indicate that further investigation in patients with small tonsils may be necessary to identify the site of obstruction prior to or in conjunction with adenotonsillectomy.

This study identifies other common sites of collapse during DISE in patients with small tonsils. We found that the most common sites of obstruction are the base of tongue and the adenoid region. Although we routinely perform adenoidectomy at the time of tonsillectomy, consideration could be given to investigating the role of lingual tonsillectomy in the treatment of children with OSA and small tonsils. In patients with small tonsils and significant OSA, DISE may be appropriate during initial surgical evaluation, even if surgical plan consists solely of adenotonsillectomy. Among patients with OSA and large tonsils, our results support first-line treatment with adenotonsillectomy. DISE likely is not needed unless there are residual symptoms of sleep-disordered breathing after surgery.

Limitations of this study include the retrospective nature of data collection regarding clinical examination findings, demographic characteristics, and risk factors for OSA. There also may be institutional differences in DISE technique. A recognized limitation of DISE is that it only serves to mimic sleep and may not accurately assess the location of obstruction or the timing of obstruction, depending on differences in technique. Because there was no way to allow for blinding of surgeons or patients regarding patient characteristics or outcomes, there is a risk of bias with scoring tonsil size or degree of LPW obstruction. We have attempted to account for these limitations by controlling for factors such as the presence of comorbid conditions in our analysis. Future studies are ongoing to address these limitations.

Additionally, we acknowledge that this study was conducted at a tertiary care institution, which likely will serve more patients with significant comorbidities and refractory OSA despite surgery. DISE may not be necessary for otherwise healthy children with at least 2+ tonsils; however, this study identifies a potential utility of DISE in patients with small tonsils or otherwise challenging aspects of upper airway obstruction.

CONCLUSION

There is a positive correlation between Brodsky Score and DISE LPW score. The majority of children with sleep-disordered breathing with a Brodsky score of 1 did not demonstrate LPW obstruction. This suggests that tonsillectomy alone may not be sufficient at treating OSA or SDB in patients with small tonsils. Further studies evaluating domains of obstruction in patients with small tonsils or comorbid conditions are necessary to help refine current practice guidelines.

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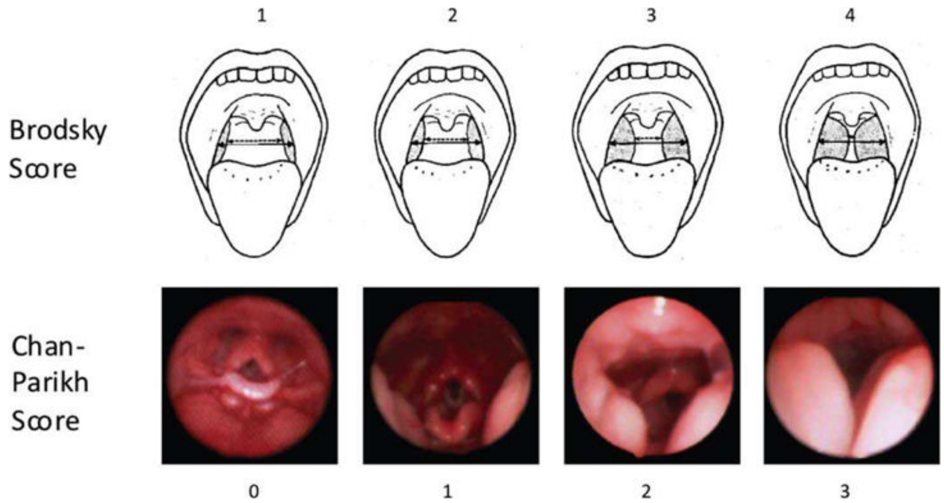

Fig. 1.

Comparison of Brodsky physical exam score and representative images of Chan-Parikh scoring for lateral pharyngeal wall domain [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

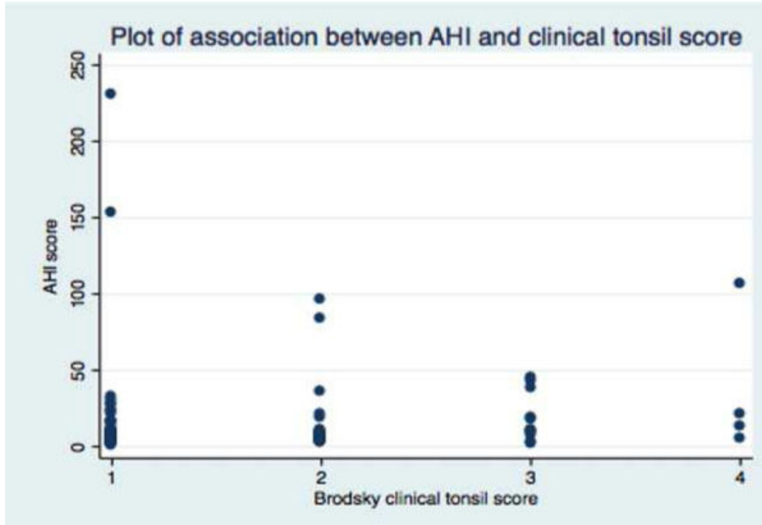

Fig. 2.

Plot of association between AHI and clinical tonsil score. AHI = Apnea-Hypopnea Index. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

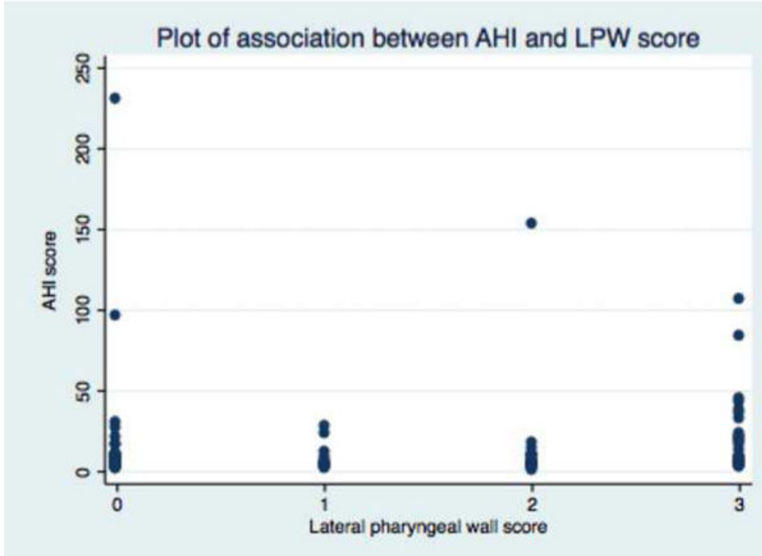

Fig. 3.

Plot of association between AHI and lateral pharyngeal wall score AHI = Apnea-Hypopnea Index; LPW = lateral pharyngeal wall.

[Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

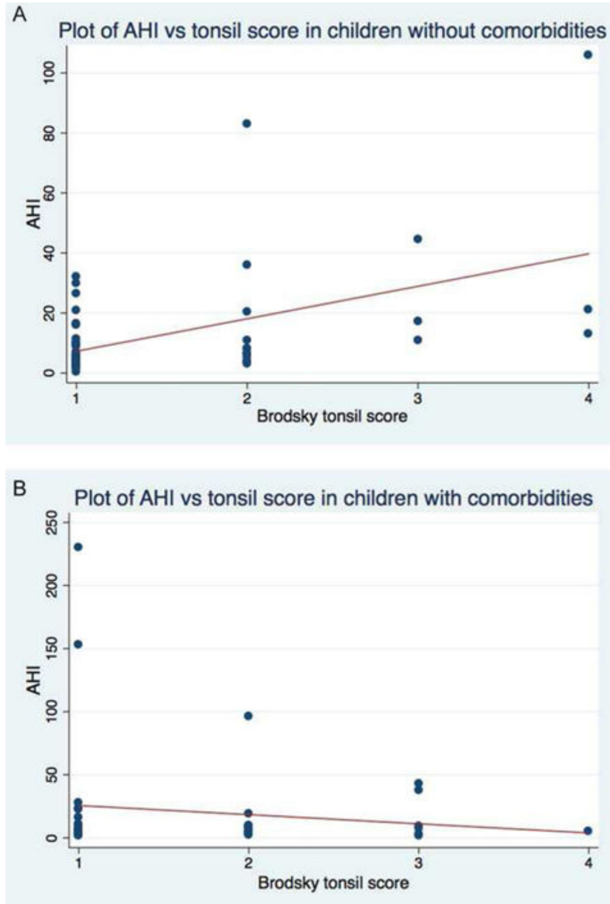

Fig. 4.

(a) Plot of AHI versus tonsil score in children without comorbidities. (b) Plot of AHI versus tonsil score in children with comorbidities. AHI = Apnea-Hypopnea Index. [Color figure can be viewed in the online issue, which is available at www.laryngo-scope.com.]

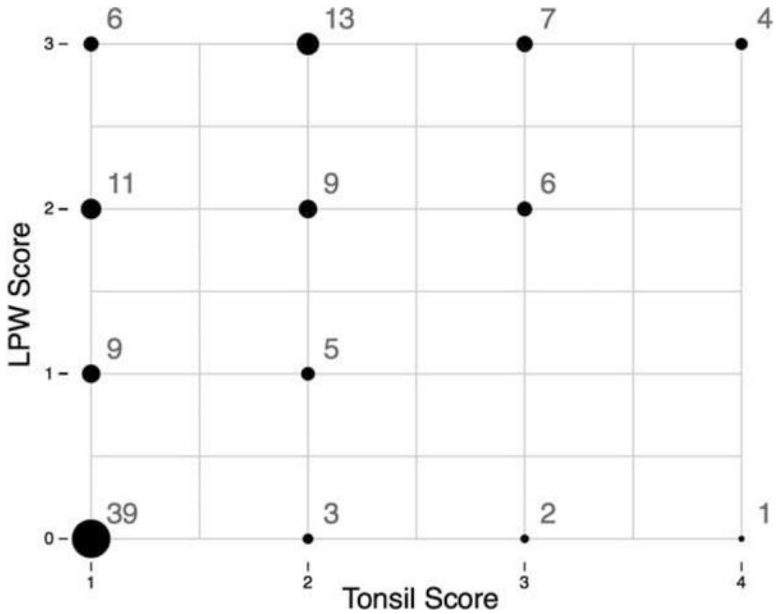

Fig. 5.

Weighted scatter plot depicting the relationship between Brodsky tonsil score and lateral pharyngeal wall score on sleep endoscopy LPW = lateral pharyngeal wall.

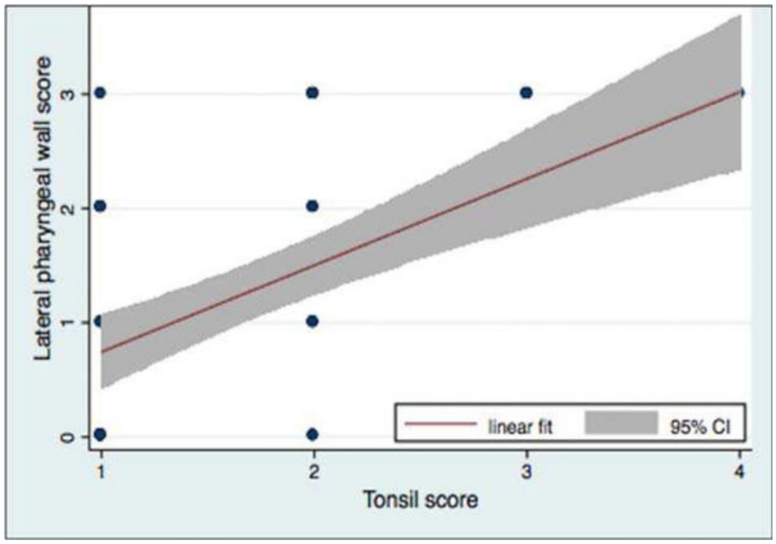

Fig. 6.

Two-way linear prediction plot with confidence interval of the association between Brodsky tonsil score and lateral pharyngeal wall score. [Color figure can be viewed in the online issue, which is available at www.laryngoscope.com.]

TABLE I

Characteristics of Patients Who Underwent DISE.

	Number	Age in Years at Time of DISE (range)	Median AHI (range)	Median O2 Nadir (range	% With Syndrome
Brodsky score					
1	65	5.9 (0.9–17.8)	5.9 (0.4–230)	90 (59–96)	37%
2	30	4.8 (0.1–18.2)	6.6 (2.2–96.3)	88.5 (59–96)	53%
3	15	4.2 (1–18.6)	11 (1.7–44.6)	89 (73–96)	67%
4	5	3.5 (2.2–14.3)	17 (5–106)	83 (68–95)	20%
Lateral pharyngeal wall score					
0	45	6.4 (0.1–17.8)	6.2 (1.7–230)	90 (71–96)	39%
1	14	5.8 (1.7–17.8)	4.8 (1.8–28)	89 (74–95)	63%
2	26	4.7 (0.9–16.2)	5.9 (0.4–11)	89.5 (59–94)	53%
3	30	4.2 (0.9–18.6)	14.7 (2.2–106)	86.5 (59–96)	41%

AHI = Apnea-Hypopnea Index; DISE = drug-induced sleep endoscopy

TABLE II

Expanded Chan-Parikh Scoring for All Patients With 1+ tonsils.

Patient	Tonsil Score (0–4)	Adenoid	Velum	LPW	Tongue Base	Supraglottis	Total C-P Score
1	1	0	0	0	3	0	3
2	1	0	0	0	0	0	0
3	1	0	1	0	3	0	4
4	1	0	0	0	3	0	3
5	1	0	0	0	2	0	2
6	1	1	0	1	2	0	4
7	1	0	0	0	2	0	2
8	1	3	0	0	0	0	3
9	1	1	0	0	2	2	5
10	1	0	1	0	3	0	4
11	1	3	0	0	0	2	5
12	1	3	0	2	3	0	8
13	1	2	0	2	0	2	6
14	1	2	0	0	1	3	6
15	1	3	3	2	3	1	12
16	1	0	3	0	3	0	6
17	1	2	2	2	0	0	6
18	1	2	2	0	0	0	4
19	1	2	2	0	1	2	7
20	1	0	3	0	1	0	4
21	1	1	0	0	2	2	5
22	1	1	0	0	3	0	4
23	1	2	0	2	0	0	4
24	1	2	0	0	0	3	5
25	1	1	0	2	2	0	5
26	1	1	0	1	2	0	4
27	1	3	3	0	0	0	6
28	1	0	0	0	3	0	3

Patient	Tonsil Score (0–4)	Adenoid	Velum	LPW	Tongue Base	Supraglottis	Total C-P Score
29	1	0	2	0	2	0	4
30	1	0	0	0	2	0	2
31	1	1	0	0	1	3	5
32	1	3	0	2	0	0	5
33	1	0	3	0	3	0	6
34	1	0	0	0	3	0	3
35	1	0	1	1	3	0	5
36	1	1	0	0	0	1	2
37	1	1	0	0	0	1	2
38	1	3	3	3	1	0	10
39	1	3	0	0	0	0	3
40	1	2	2	3	1	0	8
41	1	0	2	0	0	0	2
42	1	0	0	0	0	3	3
43	1	2	3	3	0	0	8
44	1	2	0	0	0	0	2
45	1	2	0	0	1	3	6
46	1	0	1	2	0	0	3
47	1	1	2	3	0	3	9
48	1	1	1	1	3	0	6
49	1	0	0	0	3	0	3
50	1	0	3	3	0	3	9
51	1	1	1	0	1	0	3
52	1	1	0	1	2	2	6
53	1	1	2	2	2	3	10
54	1	0	1	1	0	3	5
55	1	0	2	2	3	3	10
56	1	1	3	1	1	3	9
57	1	1	1	0	2	0	4
58	1	0	0	1	0	1	2
59	1	0	0	0	3	0	3

Patient	Tonsil Score (0–4)	Adenoid	Velum	LPW	Tongue Base	Supraglottis	Total C-P Score
60	1	0	3	0	3	2	8
61	1	1	0	2	0	3	6
62	1	0	3	0	3	3	9
63	1	0	0	0	0	2	2
64	1	1	1	1	3	0	6
65	1	2	2	3	0	3	10
	Average domain score	1.015384615	0.953846154	0.753846154	1.384615385	0.953846154	5.061538462

C-P 5 Chan-Parikh; LPW 5 lateral pharyngeal wall.